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Determining the burden of the family caregivers of people with neuromuscular diseases who use a wheelchair

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Abstract

The present study provides a basic outline of the care and support that family caregivers offer to people affected by neuromuscular diseases.

To determine the presence of burden in caregivers of people with neuromuscular diseases who use a wheelchair and to establish whether the presence of burden is influenced by contextual factors, between them, the use of wheelchair.

The applied design was cross-sectional and descriptive. The data were collected through a specific questionnaire, the Functional Independence Measure, the Matching Person and Technology form and the Zarit Burden Interview. The caregiver burden was analyzed in relation to different characteristics of the affected people, their wheelchairs, and factors related to the family caregivers themselves. The sample consisted of 41 caregivers, most of them (78.1%) being parents of the affected people.

The burden was detected in 71.7% of caregivers. The level of dependence was not related to the presence of burden. Performing care work affected carers' physical health (80.5%), their mood (68.3%), and reduced their leisure time (90.2%). The type of wheelchair and the frequency of its use were not related to the burden.

The results suggest that caregivers perceive burden, but its intensity is not related to the seriousness of the disease of the care receiver.

Abbreviations: AT = assistive technology, ATD PA = assistive technology device predisposition assessment, FIM = functional independence measure, MPT = matching personal technology, NMD = neuromuscular disorders, SD = standard deviation, ZBI = Zarit Burden Interview.

Keywords: burden of care, mobility, neuromuscular diseases, quality of life

1. Introduction

Neuromuscular diseases (NMD) refer to a heterogeneous group of diagnoses that share characteristics such as loss of muscle strength, a neurodegenerative progression, and chronicity of the pathology.[1,2] People affected by NMD may have limited functional capacity to perform certain daily activities, such as personal mobility, dressing, or grooming.[3] This situation implies an increase in demand for various support resources such as assistive technology (AT) and/or a caregiver (informal or personal assistant).[4]

Although physical treatment is an important aspect of a rehabilitation plan, the issues of accessibility and use of AT to maintain personal autonomy are arguable of the greater utility of patients with NMD, being the wheelchair the main used AT by these population.[5] A successful integration of AT into daily lives requires potential device users to explore a number of considerations: the meanings they assign to devices, their expectations of AT, the anticipated cost, and ways to understand that disability, among others.[6]

One of the immediate family members usually assumes the support for these needs arising from an NMD, the caregiver, who is the person taking responsibility for helping to cover basic and psychosocial requirements of the affected person and supervising their activities.[2]

In this context, informal caregivers can be defined as unpaid helpers, for example, family, friends, and neighbors, who assist adults with disabilities.[7]

The role of caregiver is variable and may require little involvement or a great effort, it can be a short-term commitment or lasting an indefinite period.[8] In the case of NMD, because they are chronic diseases, they usually involve a lasting experience, which requires reorganizing the family, work, and social life. In addition, the primary caregiver may be exposed to different emotions and feelings, both positive and negative.[9]

Consequently, the activities and functions performed by caregivers can have an impact on their overall health and emotional well-being. Without the appropriate precautions, they may face caregiver burden, that will be characterized by the presence of physical (back pain), psychological (anxiety, depression), or sociofamilial symptoms (social isolation, disruption of family life).[10]

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The subjective caregiver burden is a multidimensional phenomenon, and its level may be associated with a host of factors.[11] The need to provide care may affect their leisure activities, social relationships, friendships, intimacy, freedom, and emotional balance.[12]

Studies have, however, shown that support from caregiver did not only have negative effects, seeing as family members could express positive feelings and experiences, as well as a greater ability to cope with adverse situations.[11,13,14] With this in mind, one could argue that environmental factors, more specifically AT as a wheelchair, hoist, or devices for transfers, could theoretically be a facilitator and have a significant impact on the caregiver burden.

Although several studies have been conducted on the influence that different sociodemographic characteristics, health status and types of needs of people with chronic diseases have on the caregiver, few have focused on the group affected by NMD.[11,15–20]

This work has set the following objectives: to determine the presence of burden in caregivers of people with neuromuscular diseases who use a wheelchair and if this burden is associated with wheelchair's use; to establish whether the presence of caregiver burden is influenced by contextual (environmental and personal) factors, between them, the use of wheelchair, in accordance with the International Classification of Functioning.[21]

2. Methodology

2.1. Design and perspective

The study lasted 12 months and involved the application of a cross-sectional design.

It was carried out in Galicia, a region in Spain, with the collaboration of the Galician Association against Neuromuscular Diseases. This association is a nonprofit organization, made up of people affected by neuromuscular diseases, family members, and other people who provide support and guidance to those who suffer from NMD.

This study followed the code of ethics and met the criteria set by the Ethics Committee of the Medical Research Department of Galicia. All subjects completed an informed consent form before their involvement in the study.

2.2. Sample

The sample selection was carried out based on the records of people affected by a neuromuscular disease at Galician Association against Neuromuscular Diseases, because there is no official record or epidemiological study to determine the prevalence of these diseases in Galicia.

The eligibility criteria for the participants of the study were established as follows:

- Person with NMD: having a known or probable diagnosis of a neuromuscular disease; residing in Galicia; age ≥16 years;) using a wheelchair on a regular basis; needing the assistance of a caregiver to perform one or more activities of daily living. Throughout the text of the current work, authors refer to this group of participants using the terms "user" or "wheelchair user."
- Family/informal caregivers: providing support to the affected person in performing one or more activities; spending >2 hours of daily care; having a close family relationship with the wheelchair user. If one user has 2 or more caregivers, the selection of the main carer was determined to be the more number of hours (on average) spend to caregiving.

The Galician Association against Neuromuscular Diseases database was analyzed to recruit the participants. A total of 338 records were considered and the selection was performed based on the eligibility criteria. These candidates (51 people with NMD) were invited to participate in a phone call and/or email. If interested in participating in the research, an information letter and an informed consent form were sent to each of them. In the case of participants younger than 18 years old, consent was required from both the young person and his/her legal representative.

One caregiver per user was also selected, family members being those who spend more time on care work. The final sample consisted of 41 people with NMD and their caregivers (a total of 82 subjects).

2.3. Measures and variables

The studied variables concerning the person with NMD and their caregiver were selected based on findings from the review of other works on similar topics and from the same field.[17–19,22,23] The variables of the caregiver are shown in Table 1. In order to structure the study factors, these have been classified in accordance with the International Classification of Functioning.[21]

The data collection was carried out applying several tools:

- *A Specific Questionnaire*: it was elaborated by the research group to collected sociodemographical and clinical data. It was divided into 2 sections: the first one collected data from variables related to the person with NMD, including information concerning: the performance of activities and participation (and demands in terms of support needs to perform them), environmental characteristics, and information involving the features of their wheelchair. The second one incorporated several questions directed at the caregiver. It was focused on demographic characteristics, health, and social care, and needs and demands related to their care work. Those questions have been incorporated as variables because the follow-up of health status of caregiver is considered an important factor that can be influenced in quality of life of the caregiver. In our Spanish health system, the protocolized follow-up of the caregiver's health status is not well implemented. Given the significance of the present study, the questions made to the family caregiver and some question of the person with NMD are shown in Table 1.
- *Functional Independence Measure (FIM)*: it is a measurement of user's capacity, not the lack of it.[24] The questionnaire assesses 6 areas of functioning across 18 items: self-care, sphincter control, mobility, locomotion, communication, and social cognition. These areas are grouped into 2 domains: motor (13 items) and cognitive (5 items). Each item is scored using a 7-point Likert scale, ranging from 1 (total assistance is needed) to 7 (performance without any support.[25] The maximum score is 126, indicating functional independence, and the lowest score is 18, corresponding to maximum dependence. Validity, reliability, and sensitivity to change have been well established.[26] A self-report version of the FIM instrument (FIM-SR) has also been studied in people with neuromuscular disorders, showing an excellent reliability and validity.[27] FIM has been validated in Spanish.[28]
- *Matching Personal Technology Model (MPT):* MPT is a model developed for the use in the process of "matching" between the

Table 1

Specific data about the caregiver, collected in the second part of the specific questionnaire.

Sociodemographic characteristics of caregiver:	Sex: man, woman.
	Age (in years)
	Marital status: single, cohabiting, married, divorced, widowed.
	Employment/educational status: employed, part-time employed, unemployed, age-related retirement, retirement due to illness, another situation.
	Relationship with the affected person: son/daughter, father, mother, spouse/partner or extended family member.
Health and social care:	Difficulty to see a doctor and causes:
	Yes: you cannot leave the affected person unattended/problems with the time or date of appointments/or other (please specify).
	No
	What means would facilitate access to health services?
	Third-party support to care for the affected person in their absence
	Greater flexibility in scheduling appointments
	Others (please specify).
	Difficulty to attend the social service appointments
	Yes/no
	Social-health care monitoring: Do you have any scheduled appointments to identify potential health issues?
	Yes/no
Needs and demands of care tasks:	Number of hours per day devoted to the give support to your family member: hours
	Do you think your family member's disease has influenced your way of organizing your activities? yes/no
	If so, does this new organization involve planning activities well in advance? Yes/no
	Has your physical health status been affected since you have become a caregiver? Yes/no
	Have you suffered back and/or joint pain since you have become a caregiver? Yes/no
	Do you consider that support products, such as a hoist, can help you in your work? Yes/no
	Have you felt frequent changes in mood (sadness, nervousness, anxiety)? Yes/no
	Has your leisure time been reduced since you have become a caregiver? Yes/no
	Has your social life been reduced since then? Yes/no
Activities that you give support (select all applicable options)	Activities of daily life yes/no
	Homework yes/no
	Personal mobility yes/no
	Mobility in the community yes/no
	Leisure yes/no
Sociodemographical and clinical characteristics of person with NMD	Sex: man, woman.
	Age (in years)
	Marital status: single, cohabiting, married, divorced, widowed.
	Employment/educational status: employed, part-time employed, unemployed, age-related retirement, retirement due to illness, another situation.
	Diagnosis group of NMD
	Time from diagnosis: in years
	Time from the use of first wheelchair: in years
	Level of disability (>100%)
	Level of ambulation (*according Hoffman classification*): functional ambulation/nonfunctional ambulation
Activities and participation of person with NMD	Independence on ADL. *Measuring with FIM*
	Instrumental activities of daily living *(indicate those whom person is independent)*
	Education and work: *Yes/no*
	Leisure and social participation: *(indicate those whom person is independent and those for person needs support)*
Need from support of person with NMD	Type of wheelchair used: manual no self-propelled, manual self-propelled, powered.
	Other assistive devices used: *Indicate all devices used by person at this moment.*
	Numbers of hours needing caregiver:
	Activities for whose needs support from caregiver:
	Adjustments at home (done before or needed):

NMD = neuromuscular disorders.

person and the AT, taking into account the characteristics of the user, the environment, and technology.[29] The MPT instrument is a battery made up of different forms. The Survey of Technology Use helps identify technologies an individual feels comfortable or successful in using so that a new technology can be built around existing comfort or success. Highlighting the Assistive Technology Device Predisposition Assessment (ATD PA), which allows guiding decisions on important factors of use and nonuse during the selection of the most appropriate AT. The Spanish version of this instrument

has been validated.[30] From the ATD-PA, the C section has been selected to obtain a profile of the psychosocial characteristics of users, including personal motivation and perceived identity.[31–33]

- *Zarit Caregiver Burden Scale or Zarit Burden Interview (ZBI):* it is a self-administered instrument aimed at evaluating the subjective burden perceived by the primary caregiver.[19] That is why this is one of the most widely used scales in clinical practice to determine the presence of caregiver burden.[34] Burden scores are significantly associated with the quality of the relationship between the caregiver and the person receiving care,[35] the psychiatric symptoms,[36] the mood,[37] and the physical health.[38] For this research version of Zarit with 22 items has been used.[36] The ZBI has been translated and validated in Spanish,[39] and in this version, its cut-off is fixed in 47 points, so caregivers with a score of 47 or more, would have a burden. It allows divided the caregiver's sample according to their results into 2 groups: no burden (ZBI <47) and presence of burden (ZBI ≥ 47)

2.4. Procedure

The applicability of the standardized assessment tools allows these to be employed by face-to-face, telephone,[32,40–42] or self-administered interviews,[40,43,44] with precise indications. Therefore, the procedure was performed in 3 ways:

- Face-to-face interview with the users and caregivers in their home: this has been the preferred administration of the instrument, allowing a thorough understanding of the personal and environmental factors of the user, the information collected is more reliable. The duration of each visit ranged from one and a half to 2 hours, and the users and their caregivers were interviewed separately and privately.
- Telephone interview: This method was applied in the cases of researchers could not establish a home visit. Employing this one, data from 1 user and 2 caregivers were collected.
- Sending the questionnaires by mail and email: this procedure was performed in cases in which a personal interview could not be arranged or the user preferred more privacy. Questionnaires were sent, along with instructions on how to return them. This method was used to obtain data from 12 persons with NMD and their caregivers.

2.5. Analytic strategy

The quantitative variables are expressed as mean (M) $\pm$ standard deviation (SD), median and range. The qualitative variables are presented as the absolute frequency and valid percentage.

A bivariant analysis has been carried out using nonparametric methods. The correlation between the main outcome variable (subjective caregiver burden) and the other studied variables was measured using nonparametric correlations. Regarding the analysis of categorical factors, the exact Fisher test or Likelihood ratio, through contingency tables. To analyze the relationship between the presence of caregiver's burden with certain quantitative variables, these were dichotomized, taking as their cutoff the median. The significance level was set at $P < .05$ for all the hypothesis tests. All variables were analyzed using the SPSS statistical suite, in its version 18.0 for Windows, to check possible significances. In results sections, relevant data and relationships with significance were showed.

3. Results

3.1. Characteristics of wheelchair users

The users who participated in this study display a heterogeneous profile in terms of age group, diagnosis, and level of independence in activities of daily living. As seen in Table 2, most of the users are men and 80.5% of the sample is without a partner, reaching a mean age of 29.17 years (SD = 18.1).

In relation to the users' degree of independence and their need for help in performing different activities and participation, a total mean FIM score of 81.2 (SD = 17) was obtained, out of a maximum of 126. Most of the wheelchair users had a moderate level of dependence. Thus, only 1 user claimed not to need caregiver support to perform activities of daily living, but they all needed some help in carrying out instrumental or advanced activities.

3.2. Caregiver's profile

The caregivers' sociodemographic characteristics are shown in Table 3. Regarding the kindred with the user, most of the caregivers (78.1%) are their parents (in 27 cases the caregiver was the mother and in 5 the father).

Caregiver's health and social care: although only 29.3% of caregivers declared they found it difficult to see their doctor, the number of people receiving regular monitoring of their health is very low because of their caregiving obligations (3 cases). Most respondents (85.4%) perceived that if they have the presence of a professional advisor it would be positive or very positive, regarding the resources and health aspects related to the diseases

Table 2

Sociodemographic and clinical characteristics of users.

Characteristics/variables	N	%
Sex		
Male	28	68.3
Female	13	31.7
Marital status		
Single	33	80.5
Married	8	19.5
Age group		
7–17 yr	12	29.3
18–35 yr	12	29.3
36–50 yr	11	26.8
>51 yr	6	14.6
Presence of functional mobility*		
Mobile	7	17.1
Immobile	34	82.9
Diagnostic group		
Spinal muscular atrophy	5	12.2
Arthrogryposis	2	4.9
Muscular dystrophy	25	61.0
Myopathy	7	17.1
Steinert disease	1	2.4
Other	1	2.4
Age at diagnosis		
<12 yr	25	61.0
>13 yr	16	39.0
Elapsed time from diagnosis		
<20 yr	20	48.8
≥21 yr	21	51.2

* The mobility characteristics of users have been determined through the application of the Hoffer classification.

Table 3

Demographic characteristics and tasks of primary caregivers (n = 41).

Qualitative variables	N	%
Sex:		
Female	31	75.6
Male	10	24.4
Marital status:		
Single	2	4.9
Cohabiting	0	0
Married	32	78
Divorced	2	4.9
Widowed	5	12.2
Employment/educational satus:		
Employed	12	29.3
Part-time employed	2	4.9
Unemployed	6	14.6
Receiving economic support from the Spanish Government	11	26.8
Age-related retirement	10	24.4
Causes for not having a job:*		
Own decision	7	25.9
Retirement	10	37
Inability to reconcile a job and care work	8	29.6
Others	2	7.4
Affinity with the affected person:		
Son/daughter	1	2.4
Father	5	12.2
Mother	27	65.9
Spouse/partner	6	14.6
Extended family member	2	4.9
Activities in which caregiver provide support		
Activities of daily life	41	100
Homework	41	100
Personal mobility (to propel wheelchair)	10	24.4
Mobility on community	24	58.6
Leisure	25	61

Quantitative variables	Mean (SD)	Median	Range	Minimum	Maximum
Age	51.8 (14.9)	50	59	27	86
Hours per day to give care	18.7 (8)	24	22	2	24

* Percentage calculated on 27 caregivers who do not have a job.
SD = neuromuscular disorders.

that they may develop and their work as caregivers, whereas 6 of them were indifferent.

3.3. Needs and demands of care tasks

Only 1 caregiver considered that the user's disease had no influence on their health, mood, social life, or leisure time. A high percentage (80.5%) reported that being caregivers affected their physical health, whereas 68.3% suffered mood alterations and 90.2% reduced their leisure time. The presence of joint or back pain is also very common, as only 5 people (12.2%) declared they did not experience it.

3.4. Presence of caregiver burden and determining factors

Through the implementation of the Zarit Burden Interview (ZBI), it was shown that out of the 41 caregivers evaluated, only 12 (29.3%) showed no burden (ZBI < 47). When considering the mean age of the primary caregiver by level of the burden, there are hardly any differences in the data, the older caregivers (age mean = 56.3) presenting a low level of burden than younger.

Out of the caregivers exhibiting burden, low or intense,[29] the average number of hours per day devoted to the care of the family member was 18.7 (SD = 8), with a median of 24 and a range of 22 (a minimum of 2 and a maximum of 24). The mean age of these 29 caregivers is 52.8 (SD = 15), with a median of 50 and a range of 54 (a minimum of 32 and a maximum of 86). In order to do analyses finding possible influence factors of that burden, caregivers group was divided into 2 groups: those with burden (score of ZBI ≥47 points, N = 29), and those without burden (score of ZBI <47 points, N = 12).

Analyzing the possible influence of certain variables of needs and demands of giving care, it has been reported that negative perception of physical health ($P < .05$), mood alterations ($P < .05$), and reduced social life ($P < .01$) have a significant relationship with the presence of burden.

3.5. Characteristics of the people with NMD and their influence on the caregiver burden

The marital status, as a sociodemographic characteristic of the wheelchair users, had some connection with the presence of caregiver burden. In this case, caregivers providing support to married people with NMD have a lower risk of suffering from burden ($P < .05$) than those responsible for the care of single persons. A noteworthy fact is the user's sex because burden occurs more frequently in caregivers who are responsible for looking after men (n = 21) compared with those devoted to the care of women (n = 8).

No clinical feature of the person with NMD has demonstrated an influence on the occurrence of caregiver burden. In relation to the use of assistive devices, burden is more common in caregivers who do not use any AT to handle transfers (n = 14), but this fact has no significance ($P = .228$) for our study.

Certain psychosocial characteristics of the users (assessed by Assistive Technology Device Predisposition Assessment) seem to have a clear influence on caregiver perceived burden. Applying the Mann-Whitney U test to check significance with respect to different means in the ZBI score, it was found that if a person with NMD had motivation ($P < .05$), perceived identity ($P < .05$), support from family ($P < .05$), and did not feel isolated ($P < .05$), this could decrease the level of caregiver burden.

With respect to the type of activities undertaken by the person with NMD, it is worth noting that the group of caregivers who look after school-age people shows a higher level of burden than caregivers of people who are not in this situation ($P < .05$). In addition, it was found that if the people with disability carried out satisfactory activities for themselves, their carers presented a lower level of burden ($P < .01$).

The age at which the person receives their first chair and frequency of use thereof have been significantly associated with the presence of burden. In the first case, it was observed that the ZBI score has a significant inverse relationship with the age at which the persons with NMD begin to use the device ($r = -0.361$; $P < .05$). That is, the younger the person is when they begin to use their first chair, the greater the intensity of burden experienced by the caregiver. On the contrary, a higher frequency of use of the wheelchair may have a positive impact on the presence of caregiver burden ($P < .05$).

When a possible influence from the presence of architectural barriers in the house or the use of any AT to handle transfers may have on the presence of burden is analyzed, no relevant data were obtained.

Applying Spearman test, and considering as dependent variable the total score from ZBI, no significant relationship was obtained between this one and the total FIM score ($r = -0.072$; $P = .656$). In other words, the level of functional independence of the person seems to have no impact on caregiver burden.

To summarize, Table 4 shows the characteristics of the caregiver and the person with NMD for which a significant relationship was found in relation to the burden.

4. Discussion

The present study provides a basic outline of the care and support that family caregivers offer to people affected by NMD, who make use of a wheelchair. It offers a general perspective of the main characteristics of these patients, and the functions performed by their family caregivers, exploring their needs and possible burden derived from the care work. Furthermore, certain analyses were carried out to identify potential factors influencing the occurrence of burden.

This study obtained a sociodemographic profile of caregivers which, specifically in relation to sex (75.6% of them were women) and age (M = 51.8) is very similar to that obtained in previous studies on caregivers of people with NMD (mainly women and older than 50 years).[16–19,45,46]

Neuromuscular diseases are chronic conditions which may have debilitating effects on the family members of the afflicted person. Despite its importance in the quality of the received care and wellbeing, both of the caregiver and the user, this situation is a reality that is not always taken into account in the intervention process[47,48] regarding this group.

Despite the small size of the sample (41 caregivers), it is worth noting that the presence of burden was detected in 70.7% of the participants. This figure is slightly higher compared to previous studies in people with NMD (using the same burden scale), users and no users of wheelchair, in which the presence of burden slightly exceeded 50% of the population covered.[11,16,17,19] As showed in the present study, the intensity of burden may be related to the presence of physical health problems, and mood alterations and the reduced social life of the caregiver.

4.1. Caregiving and contextual factors

According to the International Classification of Functioning, contextual factors include environmental (factors that are not within the person's control, such as family, work, government agencies, laws, and cultural beliefs) and personal factors (which refer to race, sex, age, educational level, coping styles, etc). Both types of factors can be barriers or facilitators to engage a person in an activity, that is, they influence his/her functional status.[21] Consequently, it is necessary to identify which factors are barriers and which are facilitators, so that the caregivers could receive full health support and the burden related to care work could be prevented.

4.1.1. Personal factors. Previous research found that perceptions, beliefs, and attitudes are personal factors which have an impact on how an individual experiences a health condition.[49,50] Other studies also demonstrated that caregivers' attitudes and beliefs about care recipients and their health conditions affect the quality of the care provided.[51]

In the present study, contrary to what one might think, the presence or absence of burden showed no significant relationship with the caregiver's sociodemographic characteristics or with the variables of health and social care. This is different from the results obtained in other studies in persons with NMD and their families, in which a relationship was established between this burden and the social and health care support levels, the marital status,[19] and the caregiver's age.[16,17]

With respect to the number of hours devoted to care-work per day, researchers have given the opportunity to caregivers to express their level of perceived dedication, the provision of a 24-hour service being considered a full-time job. According to the obtained results, it was found that even the group of caregivers who devoted more time (hours per day) to give care than obtained average (M = 18.8; SD = 8.1) may experience a low level of burden. These data bring new evidence demonstrating that the perception of hours per day while providing care is not always associated with burden, there are other personal factors that seem to be more relevant when determining it.

On the contrary, it was shown how caregiver's health problems or alterations in their mood, since the manifestation of these issues were associated with the presence of burden. This fact confirms, in part, the findings of previous studies.[16–18,22] In order to complete these results, an empirical work should be conducted using standardized instruments which allow evaluating both caregiver's health and mood, and not only establish their relationship with the subjective burden but also estimate the direction of this association. In other words, it would be interesting to understand whether the burden is a cause or a consequence of high levels of depression and anxiety, and/or a poor perception of the health status.

In the analysis of the relationships between the variables related to the wheelchair user and the level of caregiver burden, the significant influence of certain psychosocial (motivation, perception of support) and demographic characteristics (marital status) should be noted, and the variables related to the participation in education and satisfactory activities, and the experience of wheelchair use.

The progressive nature of an NMD involves an increased dependence on the performance of daily activities and therefore an increased demand for care. This may suggest that greater dedication to this work of the family caregiver will adversely affect the perceived stress. In this sense, this influence does not seem obvious, because one of the most notable aspects, and evidence in this study, is the lack of correlation between the level of functional independence of the user (assessed through FIM) and the caregiver burden. These results are consistent with those obtained in previous studies on people with NMD and their caregivers, which also showed the absence of such a connection.[11,17,18,20,22,46] In other words, this may suggest that the stress perceived by the caregiver may not be related to the user's functional independence.

Nevertheless, our study found that there were other factors which may influence the level of burden: psychosocial characteristics of the person with NMD, the age at diagnosis, and what age wheelchair use began. Considering the above and according to Chen and Clark,[22] our conclusion is that family caregivers may have developed a specific sensitivity on coping with the daily needs of the user when identifying and anticipating the progressive nature of the disease.

The fact that the wheelchair user perceives that they have adequate support from their family and that they are not alone also have a positive effect on the absence of caregiver burden. In other words, if the user notes proper support from their family and informal caregiver, a positive feedback occurs in terms of satisfaction with their care work.

Table 4

Presence of caregiver's burden and their relationship with the characteristics of caregiver and the person with neuromuscular disorders.*

Variables		No burden (n = 12)	Burden (n = 29)	*P*	95% CI
Sociodemographic characteristics of caregiver					
Sex	Man	5	5	.106	0.119–0.132
	Woman	7	24		
Age (percentile)	<42 yr	4	10	.768	0.905–0.917
	Between 43 and 58 yr	5	9		
	>59 yr	3	10		
Hours dedicated to care	<8 h	2	5	.672	1-1
	>8 h	10	24		
Marital status	Single	1	1	.889	0.706–0.724
	Married	9	23		
	Divorced	0	2		
	Widower	2	3		
Relationship with person with NMD	Son/daughter	1	0	.086	0.072–0.082
	Mother	6	21		
	Father	1	4		
	Partner	4	2		
	Other	0	2		
Employment situation	Working	4	10	.573	0.743–0.750
	Unemployed	3	3		
	Receiving financial assistance	3	8		
	Retired	2	8		
Reasons why the caregiver doesn't work	Own decision	4	3	.130	0.371–0.390
	Retirement	2	8		
	Incompatibly	2	6		
	Others	0	3		
Health and social attention					
Difficult to visit the doctor		1	11	.059	0.066–0.076
No		11	18		
Health's follow-up by doctor		1	2	.657	1-1
No		11	27		
Difficult to visit social worker		1	6	.323	0.403–0.423
No		11	23		
Needs and demands of care tasks					
Influence of disease in family organization		11	29	.293	0.281–0.299
No		1	0		
Planning well in advance		9	27	.139	0.129–0.143
No		3	2		
Negative perception of physical health		6	27	.004	0.003–0.005
No		6	2		
Presence of back pain		9	27	.139	0.129–0.143
No		3	2		
Useful of AT in care's tasks		6	21	.155	0.266–0.284
No		6	8		
Mood alterations		5	23	.025	0.026–0.032
No		7	6		
Reduction of time to leisure		9	28	.068	0.062–0.071
No		3	1		
Reduction of social relationships		4	24	.002	0.002–0.005
No		8	5		
Sociodemographic characteristics of person with NMD					
Range of age	<18 yr	2	10	.363	0.409–0.428
	18–35 yr	5	7		
	36–50 yr	2	9		
	>59 yr	3	3		
Sex	Man	7	21	.136	0.456–0.475
	Woman	5	8		
Marital status	Single	7	26	.034	0.028–0.035
	Married	5	3		
Level of disability	<65%	0	4	.235	0.641–0.659
	>65%	12	25		
Level of ambulation	Ambulant	3	4	.431	0.641–0.659
	Not ambulant	9	25		
Diagnosis group	Spinal muscular atrophy	1	4	.329	0.361–0.380

Sociodemographic characteristics of person with NMD					
	Arthrogryposis	1	1		
	Muscular dystrophy	9	16		
	Myopathy	1	6		
	Steinert myotonic dystrophy	0	1		
	Other	0	1		
Time from diagnosis	<20 yr	5	17	.153	0.289–0.307
	≥20 yr	7	12		
Activities and participation					
Type of activities:†	Frustrating	0	1	.005	0.003–0.006
	Indifferent	1	16		
	Satisfactory	11	12		
Development of educational activity:	Yes	2	14	.049	0.078–0.089
	No	10	15		
Need support from caregiver to do activities:					
Activities of daily living (yes)	9	28	.068	0.057–0.066	
Instrumental activities of daily living (yes)	12	29	Const.	0.723–0.740	
Personal mobility (yes)	3	9	.505		
Leisure (yes)	9	24	.431	0.662–0.681	
Wheelchair					
Type of wheelchair	Manual	8	14	.234	0.311–0.329
	Powered	4	15		
Age to start to use wheelchair	≤17 yr	5	17	.259	0.489–0.509
	>17 yr	7	12		
Frequency of use of wheelchair	Daily	9	28	.034	0.057–0.066
	Weekly	3	1		
Use of AT for transfers (like a hoist)	Yes	9	18	.338	0.488–0.507
	No	3	11		

* Table 4 shows the data from those variables (as wheelchair user as caregiver). The variables which difference on average has shown a significant influence on caregiver's burden has been highlighted with red color.
† It refers to section 4 of the Survey of Technology Use from Matching Person and Technology.
NMD = neuromuscular disorders.

4.1.2. Environmental factors. Among the factors related to family structure, it is worth noting the type kindred between caregiver with the person with NMD could be influenced in the presence of caregiver's burden.[9] Nevertheless, this has not been demonstrated in the present study.

The social support is one of the main aspects that positively affects reducing the burden,[17,19] and also on achieving higher levels of quality of life.[18,22] Several studies showed that caregiver's participation in group support activities may reduce stress and improve their well-being.[52] In the present study, a reduced social life was reported by 68.3% of the participating caregivers and proved a significant relationship with the presence of burden ($P < .005$). Social isolation leads to lower levels of support[19] consequently, if the disease of the user reduced the caregiver's social life, it can also be the cause of lower social support and the occurrence of burden.

The use of a facilitating AT during the performance of transfers could improve and help in the care offered by the caregiver.[47] In this study, the presence of a hoist or another AT for transfers within the home (n = 27), however, has no significant relationship with the absence of burden ($P = .228$). Next, it would be necessary to assess whether this device is actually used or is in disuse. On the contrary, it would be relevant to determine if in the cases of caregivers who do not have an AT for transfer at the moment of study, whose use would be beneficial during transfers.

Previous studies have proven that the use of a wheelchair and the level of mobility of the user exerted an influence on the caregiver burden[16,19] and on their quality of life.[18] The current work highlights the fact that neither the type of wheelchair used by the person with NMD (manual or electric) nor the absence of functional mobility was related to the presence of burden.

However, an inverse and significant relationship was found between the ZBI score and age at which the user began using their wheelchair. This suggests that caregivers of people who began using their wheelchair in childhood or adolescence faced the most distress as a result of the use of this device by the user. A possible explanation may lie in the fact of having to accept a rapid progression of the disease of the young suffering from NMD. In addition, the use of a wheelchair for personal mobility is different from the normal development of a child, where the acquisition of motor skills allows independent progress and movement. This situation can lead to a feeling of frustration among parents. These data are related to the results obtained by Kenneson et al.[19]

Several authors determined the importance of carrying out monitoring on caregiver's social and healthcare,[16,17,53] to detect health problems that may negatively affect their welfare and care work. In this article, although no specific relationship was demonstrated between the absence of difficulties to see a doctor or the monitoring of the caregiver burden performed by the former, it is not possible to make consistent inferences in this regard. Considering the above-mentioned ideas and the fact that physical pain may have a negative impact on the relationship of provided support,[45] this aspect should be included in the care programs. The perceived health status and the presence of mood alterations, such as anxiety and depression, are strongly associated with the caregiver's stress[17] and quality of life.[15]

Although several studies have been conducted on the influence that different sociodemographic characteristics, health status, and types of needs of people with chronic diseases have on the

biopsychosocial state of the caregiver, few have focused on the group affected by NMD. Moreover, these studies have addressed specific diseases, such as Duchenne muscular dystrophy or Steinert disease, or have omitted the effect that certain devices for personal mobility, such as a wheelchair, may have on the caregiver burden.[11,15,18,19]

The limitations of this study are related to the small sample size and its cross-sectional design, which allows only detecting significant associations, but not performing a causal relationship. The sample selection could be, as well, a limitation, because participants were recruited through convenience sampling. Moreover, the obtained results refer to a specific region in Spain, with a particular social and health care service system. On the contrary, it seems obvious that NMD includes a range of heterogenic conditions, with very varying levels of severity and progression. In this case, arriving at individualized conclusions is more difficult. Therefore, extrapolation of the data to other populations must be made with caution, taking into account possible differences in welfare/aid policies.

5. Conclusions

Evidence indicates that informal care for patients with chronic diseases may involve substantial time inputs of caregivers, depending on the disease and functional status of the care receiver.[54] The completion of this work demonstrates that the majority of caregivers (70.7%) of people affected by NMD present a certain level of burden. This burden is related to the perception of health held by the caregiver, with mood alterations and reduced social life. Moreover, the degree of dependency of the user and the number of hours devoted to care work do not affect caregiver burden.

To understand the true impact of the AT, the professional must also determine its effect on caregivers. Devices can reduce the dependence of a user on the support of a third person, as well as the burden presented by family members. For this reason, the need for studies to support or refute the fact that the AT intervention decreases the demand for assistance from a caregiver becomes evident, as well as the fact that the use of these devices and their characteristics have a significant influence on the burden of this agent.

In this sense, it is essential to consider the caregiver's support as a facilitating environmental factor, and take into account their needs in planning rehabilitation programs: the process of intervention and care of people affected by NMD should also consider and involve their caregiver, because their welfare and level of perceived burden will influence the provided care work, and moreover the user's quality of life.

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